LETTER TO THE EDITOR

Dissociation between the severity of cerebrospinal fluid hypovolemia and MRI findings

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Cerebrospinal fluid (CSF) hypovolemia (spontaneous spinal CSF leak and intracranial hypotension) presents with various symptoms including orthostatic headache, neck stiffness, tinnitus, hypacusia, photophobia and nausea [1]. These symptoms worsen within fifteen minutes after sitting or standing. Meningeal enhancement is frequently observed [2]. We present a case of CSF hypovolemia with dissociation between its severity and the MRI findings.

A 38-year-old woman was admitted to our hospital because of orthostatic headache, that she became aware of five days before admission. The orthostatic headache persisted at which point she attended our hospital where the clinical manifestations indicated CSF hypovolemia. She bumped her head on the trunk of her car ten months earlier, and attended a chiropractor two months before admission. The relevance between these episodes and the disease remain unclear.

Neurological examination on admission detected no abnormalities other than the orthostatic headache. Brain MRI revealed a slight subdural hemorrhage adjacent to the falx, but there was no abnormal dural enhancement or pituitary enlargement (Fig 1-A, B). On the second day after admission, her symptoms were improved by transfusion and rest. However, her brain MRI on the fifth day after admission indicated cerebrospinal fluid hypovolemia with dural thickening, pituitary enlargement and dural sinus dilation.
(Fig1-C, D). Spinal MRI detected epidural plexus dilation, but magnetic resonance (MR) myelography could not show the leakage point of CSF. Lumbar puncture and radioisotope cisternography were not performed so as to prevent post-lumbar puncture headache. Blood examinations including the markers of collagen disorders (i.e. rheumatoid factor, MPO-ANCA, PR3-ANCA), adenosine deaminase and $\beta$-D-glucan were all normal. There were also no indications of inflammation. The symptoms were improved by transfusion and rest, and the headache disappeared after the second day. The patient was discharged on the twelfth day after admission.

After discharge there was no recurrence of symptoms, but follow up MRI at the third month still demonstrated clear dural thickening (Fig 1-E, F), demonstrating dissociation between the clinical improvement and the MRI findings. Finally, dural thickening was not detected at the fifth month follow up MRI (Fig 1-G, H).

We diagnosed this case with CSF hypovolemia because of characteristic symptoms and MRI findings. Usually, the clinical conditions and MRI abnormalities have been thought to follow a parallel course in CSF hypovolemia patients, but this case indicated that this is not necessarily so. In the early stages of this disease, abnormalities of MRI are not seen in some cases [4]. Therefore, the lack of dural thickening does not preclude the diagnosis of this condition. Although MR myelography is a useful non-invasive technique for detecting leakage lesions [5], there are some cases like ours in which the
site of leakage is unclear. There are very few reports describing the natural course of this disorder, it is possible that we have observed the common progression of this disorder. If this is indeed the natural course of this disorder then the stage with dural thickening and no symptoms may be confused with hypertrophic cranial pachymeningitis. Though the reason of the dissociation between clinical conditions and MRI findings of this case is not clear, CSF hypovolemia differs in the absence of inflammation from hypertrophic cranial pachymeningitis [3]. In addition, our case indicates that dural thickening is not necessarily related to the symptoms including orthostatic headache, and that the existence of a characteristic symptom (orthostatic hypotension) in the early stage is important in accurately diagnosing this disorder.

It is possible to significantly improve the condition of this disorder only by conservative management from the initial stage. Although it is rare to observe the natural course of this disorder, and further studies are required, it is important to take discriminative symptoms into consideration when making an early and precise diagnosis of CSF hypovolemia.
References


1.5 T Gadolinium (Gd)-enhanced T1W axial magnetic resonance images (A, C, E, G) and Gd-enhanced FLAIR coronal MR images (B, D, F, H).

On admission, a slight subdural hemorrhage adjacent to the falx was seen without abnormal dural enhancement (A, B). On the fifth day after admission, though her symptoms were dramatically improved, dural thickening appeared (arrowhead; C, D). After discharge there was no recurrence of symptoms, but follow up MRI at the third month still demonstrated clear dural thickening (E, F). Finally, the dural thickening disappeared at the fifth month follow up MRI (G, H).